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Multiceps serialis Infestation in a Baboon¹. Report of a Case Exhibiting Multiple Connective Tissue Cystic Masses.

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(Plates I & II).

INTRODUCTION.

Sporadic reports have appeared in the literature concerning the occurrence of the coenurus, *Multiceps serialis*, in human and sub-human primates. Infestation with related members of the family *Taeniidae*, the genera *Taenia* and *Echinococcus*, is fairly well known. Accordingly, a pathological analysis of a case in a monkey is of interest not only because of its "considerable potential significance in human medicine" (10), but also because of a dearth of detailed post-mortem study of the disease.

This paper is a report of a coenurus, which produced a large intra-abdominal mass together with numerous connective tissue and intermuscular cystic tumor masses in a baboon, *Theropithecus gelada*.

CASE REPORT.

The animal was a young unexhibited baboon about 4 years of age. It was born in captivity in Hanover, Germany, and was brought to this country about two to three months before death. For a few months the animal appeared to be ailing, lost weight, was noticeably weak, and had developed several small ulcerations on the dorsum of the right foot. A large mass was palpable in the abdomen. A short time before death spastic paralysis of the right lower extremity was noticed, the leg being kept flexed against the body. The animal was destroyed by its owner and the necropsy performed by us on April 27, 1939, about twenty-four hours after death.

POST-MORTEM EXAMINATION.

The body is that of a well developed, cachectic baboon (*Theropithecus gelada*) about four years of age. Moderate post-mortem rigidity is present. In each pectoral region there is a subcutaneous cystic mass, which is approximately the size and shape of a hen's egg. The one on the left is slightly larger. The overlying skin is freely movable. Similar, but smaller cystic masses are palpable in the right mastoid region, in the left axilla and over-

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lying the upper border of the left scapula. An incision over the mass in the left pectoral region reveals a well encapsulated, soft, multiloculated cystic structure lying within the subcutaneous tissues. The capsule is fairly thin and gray. Upon section, numerous small round clear cysts (bladder worms) are extruded together with some clear colorless fluid. These bladder worms measure from 0.5 to 1 centimeter in diameter. The other above-mentioned cystic masses are morphologically identical.

The lower portion of the abdomen, especially on the right side, bulges distinctly and on palpation a large mass can be felt. The right lower extremity is partially flexed at the hip and completely flexed at the knee. On the dorsum of the right foot, overlying the metatarso-phalangeal joints and also over the tarsal bones, there are several shallow ulcers with even, regular edges. The majority of these are approximately 1 centimeter in diameter and about 0.4 centimeter in depth. A similar but larger lesion is present over the medial aspect of the hallux, measuring about 4 by 1 centimeter in diameter. The bases of the ulcers are grayish and have a dry slough.

Thorax. Neither free fluid nor adhesions are present in either pleural cavity. In the left paravertebral region, just above the dome of the diaphragm, and extending over the lower four ribs, there is a firmly adherent multilocular cystic mass which measures approximately 6 by 5 centimeters in diameter. This rests upon the costovertebral junctions and portions of the adjacent ribs. At its infero-lateral margin it lies anterior and internal to another cystic mass, which is situated within the lowermost intercostal space, bulging externally, and which measures about 5 by 6 centimeters. This tumor is 2 centimeters in thickness and is located within the intercostal muscle at this site. The intra-pleural cystic mass penetrates the pleura to communicate with this intermuscular cystic mass. At no place do the cysts invade the ribs, vertebral column or spinal cord. The heart, mediastinum and diaphragm are grossly uninvolved.

Abdomen. Upon opening the peritoneum a large cystic mass presents, occupying the entire right lower quadrant, its upper pole being at the liver edge and its lower pole being at the brim of the true pelvis. It is covered anteriorly by the posterior parietal peritoneum, and is entirely encompassed by a well defined translucent capsule. Posteriorly it is firmly adherent to the muscle and connective tissue overlying the right wing of the ilium and the lower portion of the spine. The peritoneal surfaces, elsewhere, are smooth and glistening and there is no free fluid in the peritoneal cavity. The viscera have their usual position and relationships except that the right kidney and ureter and the ascending colon are displaced to the left by the cystic mass. The mesenteric nodes are similar to the pulmonary nodes described below.

Lungs. The lungs weigh 190 grams, have a smooth and glistening pleura, and retain their shape fairly well upon the table. The upper lobes are light pink in color, while the lower lobes are dark purple. Crepitaney is normal throughout. The entire surface is studded by discrete, pin-head sized, pale gray, slightly elevated nodules, which on section extend into the depth of the lung for a distance of about 1 millimeter. The largest of these occurs in the right middle lobe, measures 5 millimeters in diameter and feels firm and calcified; on section, it has a caseous core. The cut surface of the lung is grossly negative, except for the presence of small gray pin-head sized nodules which are distinctly visible throughout. The hilar and tracheo-bronchial nodes are slightly enlarged, firm and adherent to each other. On section, each presents a peripheral grayish-white thick zone, which in some is calcified, and a central cheesy-like yellowish mass.

Liver. The liver surface contains a few pin-point to pin-head sized flat grayish nodules identical with those seen in the lung. A small firm nodule, 2 millimeters in size, is likewise present. It is translucent and contains within

it two pin-point opaque yellow dots. On cut surface the hepatic architecture is regular and distinct.

The remaining abdominal viscera and the genitalia are grossly not remarkable.

Abdominal Tumor. This large oblong cystic mass measures 21 centimeters in length, 12 centimeters in width, 7 centimeters in depth and weighs 815 grams (Pl. I, Fig 1). The anterior surface is smooth and presents several well demarcated lobulations, which contain clear colorless fluid and bladder worms (larval tapeworms). On opening into the mass, it is found to be multiloculated and honeycombed due to the presence of numerous closed and intercommunicating chambers, which also contain many small and large bladder worms. The cystic mass infiltrates the underlying muscle, but does not invade the bone.

Bladder Worms. The bladder worms vary in shape, most of them being elliptical, while others are spherical or entirely irregular and branching. Many of them occur in grape-like clusters; some are pedunculated. These bladder worms possess a pale, milky-white, transparent, delicate wall, and contain clear, colorless fluid together with multiple heads, both invaginated and evaginated (Pl. II, Fig. 4A). A fully detailed description of this coenurus has been given by Schwartz (10).

Extremities: Upper. Both upper extremities show similar changes. On the medial and volar aspect of each there is a large multiloculated, well encapsulated, cystic mass, measuring approximately 12 centimeters in length and 5 centimeters in width, which extends along the entire length of the forearm, its upper pole being about 3 centimeters distal to the olecranon. It is adherent to the muscles and tendons of the forearm, and compresses and flattens them. Numerous bladder worms are visible in the loculations.

Lower: Left. Between the larger muscle groups of the posterior aspect of the thigh there is an oval, translucent, well encapsulated cystic structure, approximately 8 by 4 by 3 centimeters, which compresses, flattens, and is bound to the muscles of the adjacent region. A cyst, 3 centimeters in diameter, is present in the connective tissue of the popliteal space. A similar mass is present in the lower leg and is adherent to the muscles and tendons immediately posterior to the tibia. It displaces the gastrocnemius posteriorly, which has become a thin compressed band of muscle (Pl. I, Fig. 2, and Pl. II, Fig 3). *Right.* Cystic masses, similar in all respects to those described in the lower extremity, diffusely infiltrate the subcutaneous connective tissue and the inter-muscular fascia, producing pressure atrophy of the muscles.

The tail, on section, contains many very small cysts within the connective tissue.

The brain and spinal cord are grossly uninvolved.

MICROSCOPIC EXAMINATION.

Typical fresh miliary tubercles are found in the liver, lung and spleen. A fibro-calcific area containing lymphocytes and giant cells is present in the liver just beneath the capsule. Smaller collections of large mononuclear cells are scattered in the sinusoids; a moderate number of lymphocytes and plasma cells are found in the portal fields. In the spleen there were many plasma cells in the pulp and moderate erythro- and sidero-phagocytosis. The tracheo-bronchial and mesenteric lymph nodes show evidence of old and recent tuberculosis with calcification. The left axillary node shows caseation. Ziehl-Neelsen stain failed to reveal any acid-fast organisms. The connective tissue adjacent to the right wing of the ilium and located just beneath the lower pole of the abdominal mass shows acute and chronic non-specific inflammation with focal necrosis.

The muscle immediately adjacent to the cystic mass in the extremities reveals marked degeneration and atrophy, some edema, mild replacement fibrosis and focal collections of lymphocytes together with evidences of repair as indicated by the presence of many muscle buds.

Section of the bone (right wing of the ilium) is entirely normal. The contiguous muscle shows the changes described above. The bases of the ulcers of the right foot are formed by non-specific granulation tissue.

COMMENT.

It is to be noted that *M. serialis* infestation evokes no characteristic or specific inflammatory reaction in its sites of localization, and, indeed, very little inflammatory reaction altogether. The changes in the muscles resulting from the presence of the interfascicular cystic masses are those due to pressure.

In passing, it may be stated that calcification of tuberculous foci, as found in our case, is uncommon in monkeys.

LABORATORY DATA.

The fluid within the cysts gave a strongly positive (four plus) complement fixation reaction for *Echinococcus*. X-ray of the extremities showed cystic masses, with streaks of calcification, not invading the bone. X-ray of the abdominal mass revealed only a multicystic tumor without calcification. Post-mortem roentgenogram of the lung showed numerous calcified para-tracheal nodes. Guinea pig inoculation for tubercle bacilli was unsatisfactory. A fresh preparation of a coenurus revealed the presence of many hooklets (Pl. II, Fig. 4B).

Pathologic Diagnosis: Widespread infestation by a coenurus (*M. serialis*) resulting in multiple connective tissue and inter-muscular cystic masses located in all extremities, retroperitoneally, intra-pleurally, in the anterior and posterior chest wall, right mastoid region, and the tail. Generalized miliary tuberculosis of the lung, liver, spleen and old and recent tuberculosis of the axillary, hilar, tracheo-bronchial and mesenteric lymph nodes are present, with calcification of the latter two. Degeneration, atrophy and fibrosis of the involved muscles of the extremities. Trophic ulcers of the right foot.

DISCUSSION.

The larval cestode described above was sent to the Bureau of Animal Industry³, U. S. Department of Agriculture, where it was identified by Mr. Allen McIntosh as *Multiceps serialis theropithecii* (Schwartz, 1927).

M. serialis infestation in primates is rare. The literature on this subject has been recently reviewed by Sandground (9), who listed five cases in primates and two instances in humans. The usual intermediate host is the rabbit. References to other intermediate hosts can be found in Meggitt (6) and Hall (5). The common definitive host is the dog. Schwartz (10) described a case of *M. serialis* infestation occurring in a baboon (*T. obscurus*) with a large cystic subcutaneous tumor in the right thoracic region similar to that found in our case, and gave a careful morphologic description of the larval and adult tapeworm. Schwartz adopted the name *M. serialis* var. *theropithecii* because, although the morphology of the cestode and of the tapeworm reared from a dog was similar to, if not identical with *M. serialis*, nevertheless there was a biological difference in that he was unable to obtain

³ We are greatly indebted to Mr. Allen McIntosh for kindly identifying the bladder worm for us and to Dr. E. W. Price, Acting Chief, Zoological Division, Bureau of Animal Industry, U. S. Department of Agriculture, Washington, D. C., for his cooperation.

this cestode after feeding the adult worm to rabbits. In Scott's (11) two cases, both occurring in *Theropithecus gelada*, one animal had cystic masses in the right upper arm, submental region, pericardium, mediastinum and right perirenal tissue, while in the other, a mass in the left pleural cavity had invaded and compressed the lower dorsal spinal cord. Sandground's (9) case was atypical in location, the cyst being in the brain of a monkey (*Cercopithecus nictitans*).⁴ Railliet & Marullaz (8) reported cystic masses in the perineum of a monkey (*Macacus sinicus*) due to *M. ramosus*. Sandground (9), however, believes this is really *M. serialis*. In the two reported human cases (1, 2), one showed multiple subcutaneous tumors and the other a solitary cyst in the right buttock. In these cases, the coenurus was identified by morphological study.

According to Dévé and the Registry of the Royal Australasian College of Surgeons (both cited by Godfrey (4)), the incidence in humans of *Echinococcus* infestation involving muscle and fascia is about 5%. Generally the diagnosis of *Echinococcus* disease is made by the mere finding of cysts without a detailed morphologic study of them and by various laboratory tests, including the complement fixation, intradermal and precipitin tests. These tests, however, are now recognized to be specific for related groups rather than for individual species (3, 7). Since in our case of *M. serialis* we obtained a positive complement fixation for *Echinococcus*, we are led to consider the possibility that infestation by *M. serialis* in man may be more frequent than commonly noted. This view gains credence since as Schwartz (10) has stated: "The question of the specific identity of the coenurus stage of the tapeworm genus *Multiceps* involves primarily the number, size and shape of the hooks and incidentally the other head structures, notably the suckers and rostellum." Such an analysis is not a routine procedure in most hospital laboratories.

SUMMARY.

1. A case is reported of infestation in a baboon (*Theropithecus gelada*) by a cestode identified as *Multiceps serialis*.
2. There were numerous cystic masses in the subcutaneous and intermuscular connective tissues and a large intra-abdominal, retro-peritoneal cystic tumor.
3. This type of cestode infestation is rare in primates. So far as could be determined, this is the sixth such case to be reported in a primate. Two instances have been described in humans.
4. *M. serialis* infestation produced no specific or characteristic inflammatory response in our case.
5. An incidental finding was a fibro-caseo-calcific tuberculosis with miliary dissemination.
6. Fluid obtained from the coenurus gave a strongly positive complement fixation test for *Echinococcus*. It is suggested, therefore, that some cases thought to be "*Echinococcus*" infestation in humans, with cystic masses in connective tissue and in muscle, may actually be due to *M. serialis*.

The authors are indebted to Dr. Charles R. Schroeder for his kind cooperation; and Drs. Paul Klemperer and Sadao Otani for their valuable criticism and helpful aid in the preparation of this report.

BIBLIOGRAPHY.

1. BONNAL, G., JOYEUX, C., & BOSCH, P.
1933. Un cas de cénurose humaine dû à *Multiceps serialis* (Gervais). *Bull. Soc. Exot.*, Paris, 26, 1061.

⁴ It may be mentioned here that some authors, notably Southwell (10), believe that no sharp distinction can be made morphologically between *M. serialis* and *M. multiceps*.

2. BRUMPT, E., DUVOIR, M. E., & SAINTON, J.
1934. Un cas de cénurose humaine dû au *Coenurus serialis*. *Ann. de Parasit.*, 12, 371.
3. CULBERTSON, J. T.
1938. Recent contributions to the Immunology of Helminthic Infections. *Arch. of Path.*, 25, 256.
4. GODFREY, M. F.
1937. Hydatid Disease. *Arch. Int. Med.*, 60, 782.
5. HALL, M. C.
1919. The Adult Taenoid Cestodes of Dogs and Cats, and Related Carnivores in North America. *U. S. Natl. Mus. Proc.*, 55, 1-94.
6. MEGGITT, F. J.
1924. The Cestodes of Mammals. 282 pp. London.
7. OUTEIRINO, J.
1935. Des recherches sur la prétendue spécificité des reactions de "Ghedini-Weinberg" et de "Casoni" dans le diagnostic de l'échinococcose humaine. *Ann. de Méd.*, 38, 493.
8. RAILLIET, A., & MARULLAZ, M.
1919. Sur un Cénure nouveau du Bonnet chinois (*Macacus sinicus*). *Bull. Soc. Path. Exot.*, 12, 223.
9. SANDGROUND, J. H.
1937. On a *Coenurus* from the Brain of a Monkey. *J. Parasit.*, 23, 482.
10. SCHWARTZ, B.
1927. A Subcutaneous Tumor in a Primate Caused by Tapeworm Larvae Experimentally Reared to Maturity in Dogs. *J. Agric. Research*, 35, No. 5, 471.
11. SCOTT, H. H.
1926. Report on Deaths Occurring in the Society's Garden During the year 1925. *Proc. Zool. Soc. London*, 1, 240.
12. SOUTHWELL, T.
1930. Fauna of British India. Cestoda, Vol. II. Taylor and Francis, Fleetstreet, London.

EXPLANATION OF THE PLATES.

PLATE I.

- Fig. 1. Large intra-abdominal cystic mass, which measures 21 by 12 by 7 centimeters. Weight 815 grams. Anterior surface covered by peritoneum.
Fig. 2. Left lower limb showing cystic masses displacing muscle in thigh and leg.

PLATE II.

- Fig. 3. Left leg, higher power view, showing encapsulated cystic mass with muscular atrophy. Arrow points to muscle.
Fig. 4A. Right forearm. Cystic masses displacing muscle and showing extruded bladder worms.
Fig. 4B. Photomicrograph of bladder worm showing hooklets (fresh preparation).